

Ileo-colic intussusception with an abandoned ileal loop in a patient with a temporary terminal ileostomy - Case report and literature review

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Abstract

Introduction. Adult intussusception is very rare with an incidence of approx. 5% of all cases of intussusception and the percentage of hospitalizations due to this is 0.1%. The global incidence is 2-3 cases per 1,000,000 inhabitants a year. **Case presentation.** We present the case of a 21-year-old man patient who presented in April 2020 (during the pandemic COVID-19) at the hospital for colic abdominal pain with higher intensity in the right iliac fossa and flank, diarrhea, nausea, vomiting, abdominal flatulence. Heredo-collateral antecedents are insignificant and in personal antecedents show acute appendicitis in 2007, ileal loop volvulus in March 2020 with necrosis on the adhesive flange for which enterectomy was performed with the abandonment of the downstream loop and ileostomy (surgery was performed in another medical unit). The ultrasound examination found the presence in the right iliac fossa and right flank of a small bowel loop with thickened wall, doubled, aperistaltic with hypoechoic wall and tortuous appearance being a high suspicion of intestinal intussusception and at the computer-tomographic examination the suspicion of ileocolic intussusception is confirmed. Laboratory tests showed: leukocytosis and a slight increase in CRP. Surgery was performed and ileocolic intussusception was found on the terminal ileal loop abandoned with compression on the ileostomy loop and the removal of the ileostomy, straight hemicolectomy with ileo-transverse-anastomosis was performed. The postoperative evolution was simple, being discharged at 10 days. **Discussions.** Intestinal intussusception was first described in 1674 by Paul Barbette, and the first intussusception surgery was performed in 1871 by Sir Jonathan Hutchinson. The classification of intussusceptions is made according to several criteria being classified in: anatomical location - entero-enteral, ileo-cecal, ileo-colic, and colo-colic, and depending on the cause it can be benign, malignant, or idiopathic. It is also classified in intussusception with/without triggering factor. The clinic of intussusception is extremely varied depending on the cause. Small bowel intussusception with/without cause is the most common form. The most common factors that cause this type of intussusception are the Meckel's diverticulum, certain protruding malformations of the veins, inflammatory polypoid formations, lymphomas, metastatic malignant fibrous histiocytomas. Large intestinal intussusception is caused in over 50% of cases of primary or secondary malignant lesions; the ileo-cecal and ileo-colic ones most often cause a metastatic malignant lesion with malignant melanoma as a starting point, while the colo-colic type is most commonly caused by lipomas. **Treatment.** The intussusception treatment is performed immediately: in children, attempts are made to reduce the intussusception by barium or air enema (with the pediatric surgeon in the radiology room), and if it is an adult surgery is performed, the enema is used extremely rarely and with little effectiveness. **Conclusions.** Intestinal intussusception in adults has a very low incidence. Finding the cause is not an end in itself, adult intussusception being the prerogative of surgery, the vast majority of which are solved only by surgery. In the case of children, the situation is different, with conservative methods taking precedence over surgical ones.

Key words: ileo-colic intussusception, abandoned ileal loop, ileostomy

Introduction

The adult intussusception is a very rare clinical situation being accredited with approx. 5% of all cases of intussusception at all ages and approx. 1-5% of all cases of intestinal occlusion [1]. Intussusception consists of telescoping a proximal segment of the gastrointestinal tract into the lumen of the underlying segment [2]. Ileo-

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colic invagination is even rarer, and such a clinical case is exceptionally common (we have not found any similar case in the literature). Also, the percentage of hospitalizations caused by an invagination is 0.1% of the total. Adult intussusception is a very rare pathology with an average global incidence of 2–3 cases per 1,000,000 inhabitants in a year [3].

Intussusception is a major surgical emergency, the surgical intervention being necessary to be performed as soon as possible within 0–6 hours.

Case report

We present the case of a 21-year-old man from rural areas, without occupation, who presented in April 2020 (during the pandemic COVID-19) at the hospital's surgery office of the hospital being brought by ambulance for intense colic abdominal pain, diffusely distributed in the abdomen, with greater intensity in the right iliac fossa and right flank to the right hypochondrium, accompanied by diarrhea, nausea, vomiting, abdominal flatulence. Regarding the heredo-collateral antecedents, nothing pathological is found. Regarding the personal antecedents, it is noted that in 2007 he presented acute appendicitis for which appendicectomy was performed, and in March 2020 ileal loop volvulus with necrosis on the adhesive flange for which enterectomy was performed with the abandonment of the loop downstream of the resected portion and externalization of the upstream portion in ileostomy (surgery was performed in another medical unit).

The general examination showed slightly pale skin and mucous membranes, tachycardia, weathered abdomen, with signs of peritoneal irritation, intense pain that diffuses spontaneously and on palpation in orthostatism, symptoms that decrease greatly in intensity in clinostatism; impalpable liver and spleen, intestinal transit absent for gas and faeces by ileostomy in orthostatism and partially resumed in clinostatic position. On rectal examination, aqueous stools are noticed, with

Clostridium difficile enterocolitis being suspected, which has been refuted (history of hospitalization in the last 14 days).

The ultrasound examination found the presence in the right iliac fossa and right flank of a small bowel loop with thickened wall, doubled up to 24mm, aperistaltic with the hypoechoic wall, and tortuous appearance being a high suspicion of intestinal invagination with possible necrosis of the wall [figure 1].

On abdominal-pelvic computed tomography examination, the presence of a thick-walled intestinal loop with a thickened wall, doubled up to 20 mm, with gas bubbles inside, with a tortuous path, up to subhepatic appearance suggestive of intussusception [figure 2].

Laboratory tests showed: leukocytosis (15900/mm³) and a slight increase in CRP (3.947mg/dL).

Surgery was performed and on exploration, an ileocolic invagination was found on an abandoned terminal ileal loop with compression on the ileostomy loop. An attempt was made to disinvaginate the loop but it was not successful and consequently, the abolition of the ileostomy was performed, with straight hemicolectomy with latero-lateral ileo-transverso-anastomosis, drainage with a tube [figure 3, 4, 5]



Figure 1. Ultrasound appearance of the invagination tumor.

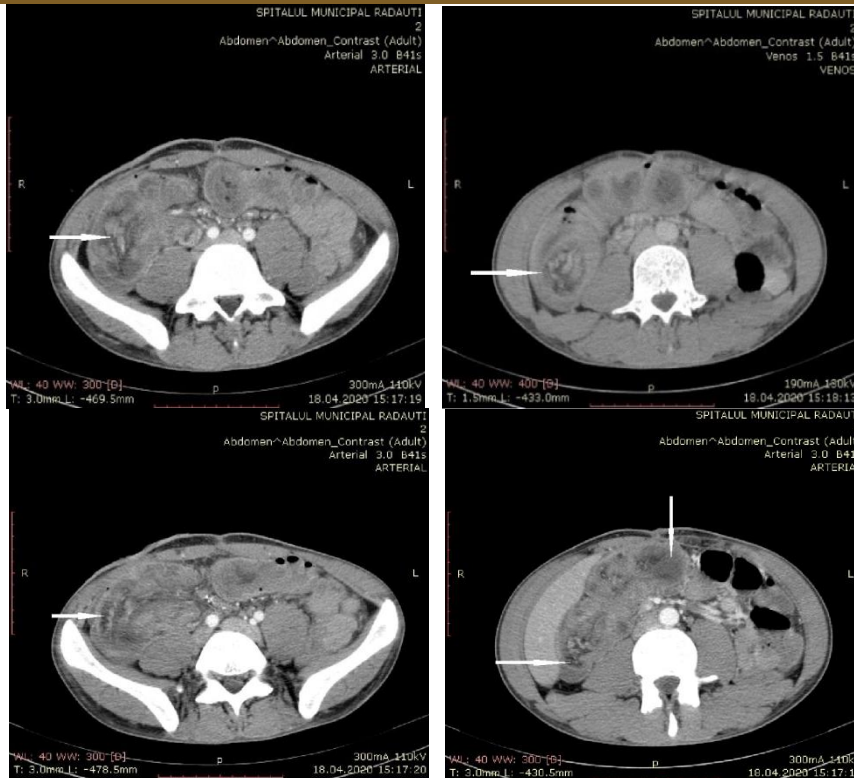


Figure 2. Presence of invagination tumor occupying the iliac fossa, right flank, hepatic angle and right 1/3 of the transverse colon.

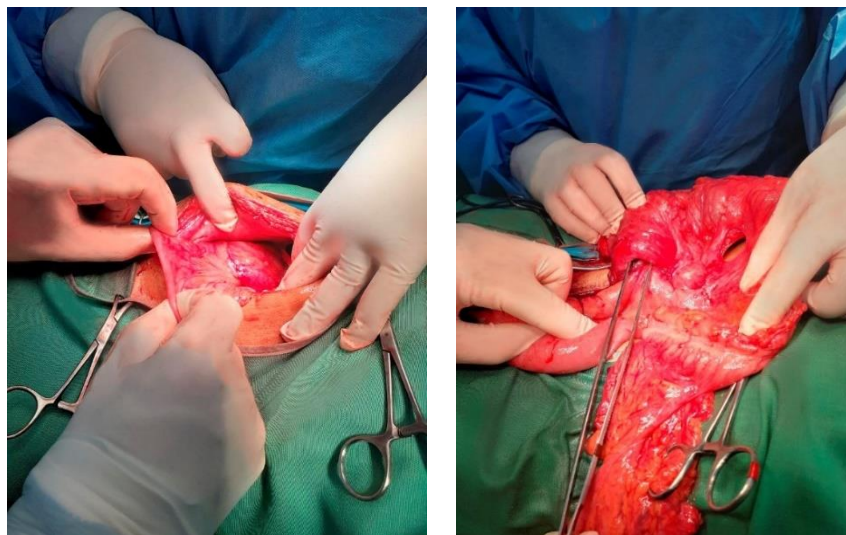


Figure 3. a - The compressive effect of the intussusception on the ileostomy;
b. The invagination orifice.

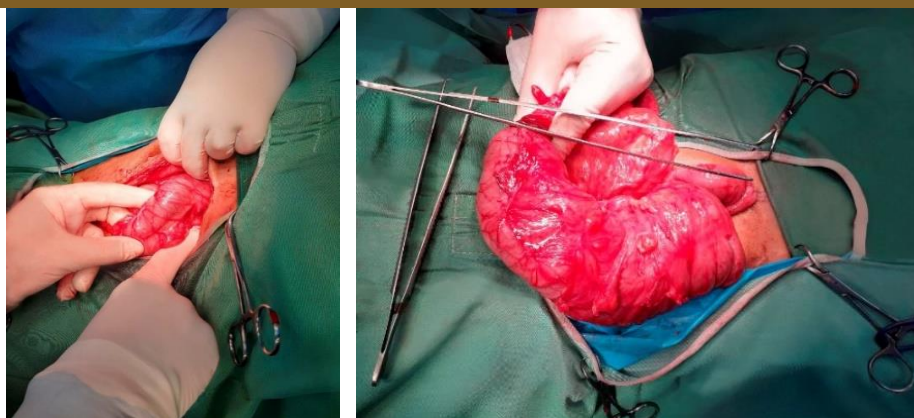
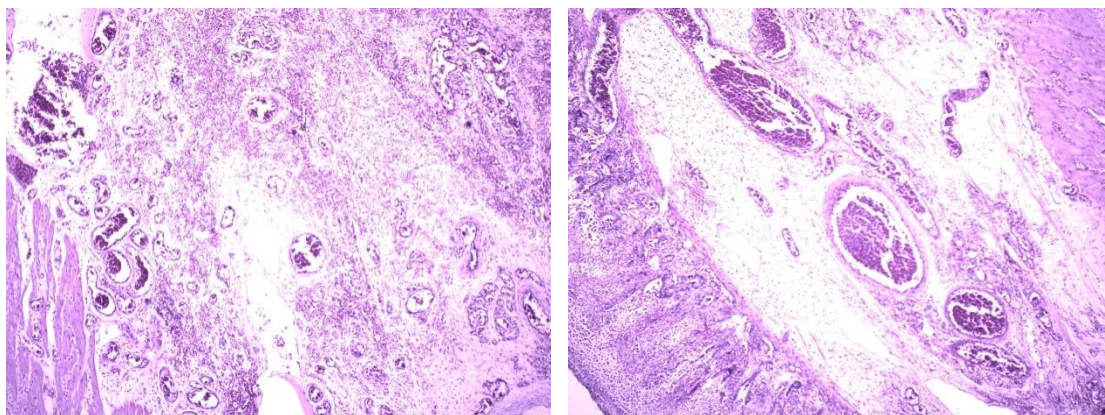


Figure 4. a. - The limit of intussusception progression in the transverse colon;
b - Resection limit at the level of the transverse colon.



Figure 5. a. Right hemicolectomy piece with intestinal invaginated loop; b. Appearance of the right hemicolectomy piece.

The histopathological examination confirms the presence of intestinal infarction with interest in the wall entirely and mesocolon [figure 6].



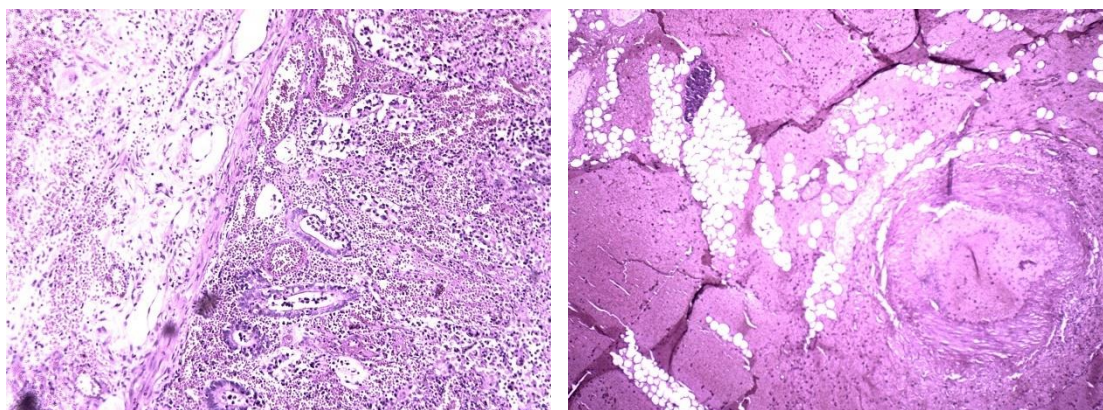


Figure 6. a. Mucosa with ischemic necrosis with blood infiltrate, vascular thrombosis and rare leukocytes; b, c. Entire intestinal wall with ischemic necrosis in the mucosa and muscle with blood infiltrate, vascular thrombosis throughout the wall and rare leukocytes; d. Mesocolon with vascular thrombosis and areas of hemorrhage.

The postoperative evolution was simple with the resumption of transit for gas at 48 hours, and for faeces at 72 hours, the duration of hospitalization being 10 days when he was discharged in a very good general condition, with normal food intake, normal transit and diuresis, and both wounds are normal.

Discussions

Intestinal intussusception was first described in 1674 by the Dutch physician Paul Barbette [4] who proposed gastrotomy as a therapeutic solution. The first successful intussusception surgery was performed in 1871 on a child by Sir Jonathan Hutchinson [5]. Intestinal intussusception is rare in adults, the percentage being 5% of all cases. Approximately 90% of cases involve the small intestine and large intestine, the remaining 10% being located in the stomach or stomas [5]. In the case of children, the factors that cause the onset of intussusception are usually unknown [6].

Risk factors for intussusception are:

1. sex - the men are the most affected;
2. age - it is well known that approx. 90% of cases of invagination involve children and are the most common cause of occlusion between 6 months and 6 years [6];
3. Also, intestinal malformations, in particular disorders of rotation and development, and particular attachment disorders, may present a risk of developing invagination;
4. In terms of history, it has been observed that siblings of a patient who has had an invagination have a higher risk of developing this condition, and people who have had an intestinal invagination in their personal history have an increased risk of having it again [6].

The classification of intussusceptions is made according to several criteria: depending on the telescopic segment it can be: antegrade when the telescopic segment is proximal or retrograde when the telescopic segment is distal [7] or depending on the anatomical location in entero-enteral, ileo-colic, ileo-cecal and acolo-colic or depending on the cause in: benign, malignant or idiopathic [8]. Depending on the triggering factor of the invagination, it can also be included in another classification intussusception with/without [8].

In the last 10 years, I have met and operated on adults 4 cases with 3 types of invagination: ileo-ileal

invagination with triggering factor a 4 cm pedicle polyp being needed segmental enterectomy with side to side anastomosis and another caused by the diverticulum Meckel [9], jejuno-gastric invagination at 58 years of age after gastric resection 2/3 in which only one episode of vomiting was found, the patient refused surgery and in this case it was an ileocolic invagination on the abandoned terminal ileal loop.

What I noticed about the 3 types of invaginations are the following: ileo-ileal invaginations were presented at the hospital several times to investigate an anemic syndrome by performing endoscopy and colonoscopy, cataloging deficiency anemia, and then the diagnosis was made. Either established by abdominal CT; jejuno-gastric invagination presented to the hospital as acute abdomen, the ultrasound examination raising the suspicion of invagination subsequently confirmed by CT examination [10]; ileocolic invagination was presented as an alternating occlusive syndrome, the diagnosis is established by CT examination.

Intussusception without a trigger can be manifested by vague abdominal pain without other symptoms. Very rarely it can cause intestinal obstruction. Most cases are discovered incidentally during a CT scan performed for other reasons [5]. All structures participating in the realization of the invagination boudin (intestinal wall, mesenteric and vascular fat, returning wall, intraluminal space) are perfectly visible on CT examination, the presence of the intestine in the intestine $\pm$ vessels or mesenteric fat being pathognomonic for invagination.

Intussusception with a trigger is the form found in adults and it is identified in 90% of the cases [11, 12]. Exceptionally, the cause that determined its appearance cannot be found. The factors that can determine the appearance of invagination are very varied (table 1).

The clinical picture of the intussusceptions that present a cause is extremely varied. Colic pain accompanied by nausea and vomiting dominate the clinical picture suggesting intestinal occlusion. If the triggering factor is a neoplastic formation then the symptoms related to malignancy are in the foreground: constipation, weight loss, melena, rectal bleeding, the presence of an abdominal tumor, the symptoms related to invagination taking second place. In the present case,

abandoned terminal ileal loop was telescoped after resection of the infarcted loop at the previous operation with the mesenteric fold in the right colon due peristalsis. Thus formed a sign considered pathognomonic for invagination and which is produced by the presence of intussusception tumor and not found in other conditions: cylindrical appearance with 3 concentric rings similar to a shooting target

Table 1. Differences between adult and childhood intussusceptions (5)

	Children	Adults
Percentage of all intestinal intussusceptions	95	5
The cause of intestinal obstruction	Frequently	Rare (5% of all intestinal obstructions)
Ethiology	90% - unknown (Peyer's plate enlargement is thought to be the trigger in almost 50% of cases)	Rarely idiopathic. The cause is identified in 70% –90%
Clinic (clasic triad: vomiting, rectal bleeding and abdominal pain)	Ordinary present	Appears in only 15% –20%
Treatment	In most cases dysfunctional (barium enema reduction) Surgical treatment is required	Surgical resection almost always

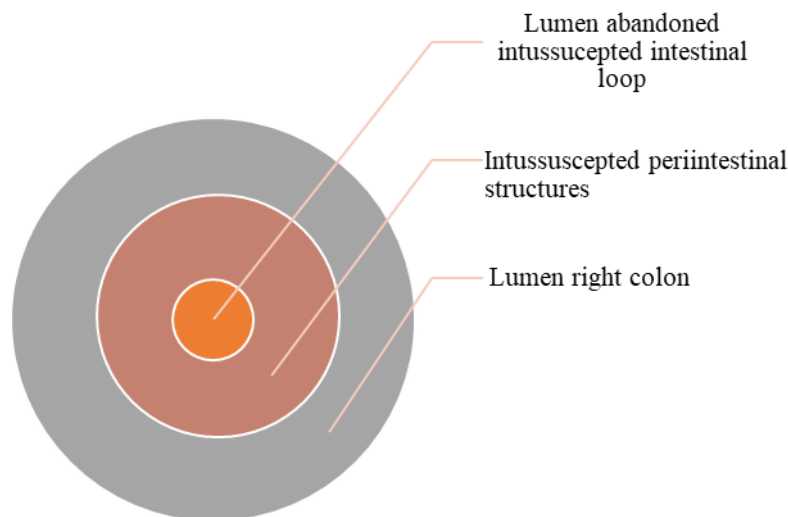


Figure 7. Schematic representation of the ileo-colic intussusception of the abandoned terminal intestinal loop.

Identifying the triggering factor of invagination is the great test for the radiologist and it is the one that practically establishes the therapeutic attitude. What also

helps us in establishing the surgical behaviour is the age at which it occurs: children develop invaginations without triggering factors and are treated conservatively in most cases, while adults make invaginations triggered by a

causative factor, and the treatment is only surgical. That is why in the face of an invagination depending on the age at which it appears, we have a certain diagnostic assumption and we base it on a certain investigative logic.

A. Intussusception of the idiopathic small intestine is the most common form and occurs as a non-obstructive portion of smaller caliber and shorter length compared to that at which a cause is found [2, 8, 9, 10, 13]. The one in which a triggering factor is identified and in which the small intestine is involved has as a cause a benign condition, but it can be, much less often, a malignant lesion, usually metastatic. Those that are located in the colon are usually caused by primary or metastatic neoplasms. In general, if the factor that triggered the invagination is identified or the invaginated segment is long, has a relatively large diameter and causes upstream dilation, surgery is required [8, 9, 10, 13]. Often they are found and diagnosed during abdominal examinations for atypical pain and for which recourse was made to computed tomography, enteroclysis, enteroclysis-CT, examination with video capsule [8].

The most common factors that can trigger a small bowel invagination are Meckel's diverticulum, certain protruding vein malformations, inflammatory polypoid formations, lymphomas, metastatic malignant fibrous histiocytomas [8].

1. Meckel's diverticulum is the most common congenital anomaly of the gastrointestinal tract, and occurs in 2% -3% of the population [2, 8, 9, 10, 13]. Complications of the diverticulum can be multiple but the most common are bleeding, small bowel occlusion and diverticulitis [8]. A relatively rare but very possible complication is small intestinal invagination triggered by Meckel diverticulum reversal. At the time of diverticulum, a reversal occurred the invagination is primed and the length depends on how strong the peristaltic waves are and the interval elapsed until the surgical intervention [9, 10, 13]. The recognition of this event in CT is difficult and it is extremely important for the speed of surgery, and finding signs of CT for diverticular reversal allows the diagnosis to be made prospectively, the intervention can be performed very early without significant visceral damage [8].

2. Inflammatory polyp of the gastrointestinal tract is rare in the small intestine. In fact, it is an inflammatory pseudotumor-like lesion or myofibroblastic tumor that usually occurs in the stomach, followed by the small intestine. Inflammatory fibrous polyp rarely causes invagination [2, 8].

3. Venous malformations in the gastrointestinal tract are externalized by bleeding. The most common place to find it is the cecum, then the right colon and jejunum. They usually occur in elderly patients with a history of cardiovascular disease. In young patients, they are found exceptionally in atypical places, including the small intestine [8].

4. Primary gastrointestinal lymphomas represent between 20-40% of all small bowel malignancies the 2 forms that meet with T cells and B cells developing in different places and somewhat in the first phase manifesting differently. T-cell lymphomas primarily affect the proximal intestine, and B-cell lymphomas are of particular interest to the distal ileum. The sudden onset of pain in a known patient with intestinal lymphoma should make us consider a possible invagination. Computed tomography is the investigation of choice used to evaluate patients with

gastrointestinal lymphomas, thus being able to evaluate both intestinal and extraintestinal components. Regional or mesenteric lymphadenopathy associated with the presence of intestinal tumor masses may represent the presence of lymphoma that may underlie an invagination [2, 8].

5. Metastatic malignant fibrous histiocytoma may be an unusual cause for intestinal intussusception when intraluminal tumor mass is manifested that may initiate the progression of an intestinal telescoping. It generally affects adults over the age of 65, and affects the extremities, trunk, retroperitoneum, and very rarely the visceral organs [14, 15].

B. Intussusception of the large intestine is caused by more than 50% by the presence of a primary or secondary malignant lesion [16]. Other causes that could be involved in triggering the invagination are colic lipomas, adenomatous polyps, and the clinical manifestations are dominated by the colic pains that determine the patient to address the medical unit. The diagnosis is generally established imaging. Transient colonic invaginations have also been described [17].

C. Ileo-cecal and ileo-colic intussusceptions are caused by a metastatic malignant lesion, the most common being a malignant melanoma with a frequency of 2-5% of all patients with melanoma. These invaginations caused by secondary lesions with melanoma starting point are very rare [18]. The only treatment used in these patients is immediate emergency surgery [19].

D. Colo-colic intussusceptions are caused by benign tumors including lipomas, adenomatous polyps, and mesenchymal tumors, and by malignant tumors including colic adenocarcinomas. Lipomas are the most common benign lesions that can trigger colonic invagination [20]. Colon lipomas originate in the submucosa in 90% of cases, are usually solitary and can be sessile or peduncled. In addition to symptoms, they are found incidentally on endoscopic or radiological examination and can be easily diagnosed on CT examination [7, 8]. The appearance of pain in the case of a lipoma should make us think of an invagination and immediately request an abdominal CT. Among malignant tumors, adenocarcinomas are the most common colonic neoplasms that can cause colo-colic invagination [8]. The clinical picture of neoplasia dominates the symptoms, and the symptoms of invagination take a back seat. If the suspicion of invagination is raised, the first recommended investigation should be abdominal CT with contrast. The diagnosis is generally easy, as the layers of the colic wall can be seen clearly.

Treatment

Treatment of invagination is performed immediately. The timing of the intervention involves major risks that can lead to severe complications such as dehydration, severe shock, and in the case of intestinal infarction increased risk of infections [6]. After a short period of hydroelectrolytic rebalancing, the evacuation of the gastrointestinal tube (nasogastric tube) in children is tried to reduce by barium enema the invagination (with the pediatric surgeon in the radiology room), and if it is adult surgical intervention, the enema being used extremely rarely and with little effectiveness. As the first step after intraperitoneal exploration, an attempt will be made to disinvagination, which in adults has little chance of success, most often requiring the resection of that portion [8]. If it is possible to safely disinvagination, then it is tried to perform it, and if the perforation of the invaginated segment or its

infarction is suspected, no conservative treatment is tried, the surgical gesture being the saving one.

The case presented is special due to the age at which it appeared and the cause that determined the appearance of the invagination. Also, the method of treatment is important, the disinvagination attempt not being successful primarily due to the time interval from the onset of symptoms to the surgical resolution of the condition.

Conclusions

Intestinal intussusception in adults has a very low incidence. Finding the cause is not an end in itself, adult intussusception being the prerogative of surgery, the vast majority of them being solved only by surgery. In the case of children, the situation is different, with conservative methods taking precedence over surgical ones.

Conflict of Interest

Authors have no conflict of interest to disclose

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